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Effect of expiratory muscle strength training on voice and speech: An exploratory study in persons with Parkinson's disease or multiple sclerosis

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Abstract

Purpose: This study explored how respiration, voice, and speech were affected following expiratory muscle strength training (EMST) and maintenance training in persons with Parkinson's disease (PD) or multiple sclerosis (MS).

Method: Nine participants with PD and six with MS participated in a randomised study, where the effects of EMST, sham, and maintenance treatment were investigated. Outcome measures included maximum expiratory pressure (MEP); maximum phonation time (MPT); intelligibility; verbal diadochokinesis (DDK); speech rate; a self-report form on voice, speech, and communication; and open questions about how the participants experienced the intervention. Group comparisons were performed within and between groups.

Result: The PD and the MS groups both improved significantly in MEP, and this improvement remained after 3 months of maintenance EMST. An improvement was also seen in DDK. Post-EMST, 33% of the PD group and 80% of the MS group reported a positive effect on communication.

Conclusion: The results of this study support previous evidence that EMST has positive effects on expiratory pressure in persons with PD or MS, but its effect on voice and speech remains unclear. Since subjective reports of the intervention and effects on communication were predominantly positive, further research is needed on larger groups to explore appropriate outcome measures.

Keywords: *Parkinson's disease; multiple sclerosis; voice; speech; expiratory muscle strength training; treatment*

Introduction

Respiratory dysfunction is a common consequence of progressive neurodegenerative conditions. Parkinson's disease (PD) and multiple sclerosis (MS) are two of the most common neurodegenerative diseases affecting the central nervous system. Despite differences in aetiology and disease progression, both diseases often affect respiratory function which, in advanced stages, can cause morbidity and mortality (Aboussouan, 2005; Gosselink et al., 1999). Exactly what mechanisms cause the disorders in the respiratory system is not fully known in PD, but it is likely that they are consequences of several factors including neurodegeneration affecting respiration at the central level, stiffness of thorax, and weakness of respiratory muscles (Pokusa et al., 2020). In MS, demyelination and axonal loss in the central nervous system may result in muscle weakness, including respiratory muscles (Aboussouan, 2005; Tzelepis & McCool, 2015; Gosselink et al., 1999), and there is evidence that weakness of the expiratory muscles is present also in

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the early stages of MS (Chiara et al., 2007). Respiratory dysfunction linked to voice and speech production is common in both PD (Darling-White & Huber, 2017) and MS (Noffs et al., 2018).

Both diseases develop gradually with increasing motor, sensory, and cognitive problems to varying degrees. Motor speech involvement in PD usually manifests as hypokinetic dysarthria, characterised by monotonic pitch and loudness, irregular rate with rushes of speech, imprecise articulation, and reduced stress (Duffy, 2019). The dysarthria in MS is often a mixed spastic-ataxic dysarthria characterised by slowness, an increased length and number of pauses, imprecise consonants, weak or strained voice, and effected pitch and loudness control (Noffs et al., 2018). Dysarthria has serious consequences when it comes to the individual's ability to make themselves understood and participate in professional and social life. About 90% of individuals diagnosed with PD state that speech and communication have changed as a result of the disease (Schalling et al., 2017; Miller, 2017), whereas about 30–50% of individuals diagnosed with MS report speech and communication difficulties (Hartelius et al., 2000; Johansson et al., 2021). Exploring treatment options that can have a positive effect on speech and communication is of great importance in both PD and MS.

Weakness and dysfunctional neural control of the expiratory muscles can have a negative impact on voice and speech due to inadequate respiratory pressure for voice production (Laciuga et al., 2014). Expiratory muscle strength training (EMST) is an intervention that aims to improve expiratory muscle strength using a resistive or pressure threshold device. Protocols for EMST vary in intensity, but a commonly used protocol is five sets of five forceful exhalations into the threshold device, 5 days a week for at least 5 weeks. In threshold devices, the threshold pressure usually ranges between 30–75% of the person's maximum expiratory pressure (MEP; Laciuga et al., 2014). Positive effects on respiration in terms of improved MEP have been reported in both PD (Darling-White & Huber, 2017; Kuo et al., 2017; Reyes et al., 2020; Sapienza et al., 2011) and MS (Chiara et al., 2007; Gosselink et al., 2000; Silverman et al. 2017; Smeltzer et al., 1996; Srp et al., 2021), as well as other patient populations with reduced expiratory muscle strength (Desjardins & Bonilha, 2020; Laciuga et al., 2014). Besides improved MEP, studies evaluating EMST have seen effects on cough function (e.g. Pitts et al., 2009), dysphagia (e.g. Claus et al., 2021; Silverman et al., 2017), and aspects of quality of life (e.g. Kuo et al., 2017). A few studies have included a detraining phase, i.e. a period of no training, followed by a new assessment (Chiara et al., 2007; Claus et al., 2021; Srp et al., 2021; Troche et al., 2014). These studies have found a decline (often small) in MEP after a period of no training, suggesting that there might be a need for a maintenance program or booster sessions in order to sustain the improvement yielded by EMST (Srp et al., 2021; Troche et al., 2014; van de Wetering-van Dongen et al., 2020). To our knowledge, no study has yet evaluated the effect of such a maintenance program.

Although there is growing evidence that EMST has positive effects on expiratory pressure in persons with neurodegenerative diseases such as PD and MS, the effects on voice and speech vary between studies and have hitherto remained quite small (Desjardins & Bonilha, 2020; Levy et al., 2018). Chiara and colleagues (2007) were the first who investigated the effects of EMST on voice and speech in patients with neuromuscular disease. In their study in persons with MS, no effect was seen on maximum phonation time or speech rate directly after 8 weeks of EMST. However, both measures had improved after a 4 week long detraining (i.e. no training) which the authors discuss might be due to a learning effect. No change was seen in the voice-related quality of life, but the participants with MS reported significantly less impact of dysarthria post-EMST. In a single-subject design study by Johansson et al. (2013), five persons with MS underwent 6 weeks of EMST. The results of that study showed promising effects on voice and speech, since all five increased their voice intensity. Three out of five participants increased their maximum phonation time, four out of five increased their intensity during maximum phonation time, and three out of five increased their intensity during reading. Studies exploring the effects of EMST on voice and speech in persons with PD are also scarce. In a study by Reyes et al. (2020) the effects of inspiratory or expiratory muscle training on voice production in persons with PD were evaluated in a randomised control trial (RCT). The study showed that expiratory muscle training had an effect on subglottic pressure (measured in plosives), maximum phonation time, and voice intensity. Darling-White & Huber (2017), on the other hand, did not find consistent changes in phonation and speech (voice intensity and utterance length) post-EMST in their single-subject study with 12 participants that aimed to improve speech breathing in persons with PD.

In summary, previous studies evaluating the effects of EMST in persons with MS or PD found positive effects on respiration, but several studies emphasise the need to implement a maintenance program to sustain function following EMST. The findings of the effect on voice and speech are mixed and limited in both groups. It is possible that effects on voice and speech take longer to implement, and studies investigating the impact on voice and speech requires a longer follow-up. Hence, the purpose of the present study was to evaluate the effects of EMST on respiration, speech, and voice after 5 weeks of EMST and

then again after 12 weeks of maintenance EMST. The specific research questions were:

- What are the effects of EMST on maximum expiratory pressure (MEP) and maximum phonation time (primary outcome measures), verbal diadochokinesis, intelligibility, and speech rate (secondary outcome measures) in persons with MS or PD?
- What are the effects of EMST on the degree of self-perceived difficulties concerning speech, voice, and communication for the participants with MS or PD, and how did the participants experience the intervention?

Method

Study design

This was a randomised trial where the participants were randomly assigned to begin with either EMST treatment (Group A) or sham treatment (Group B), i.e. similar treatment protocol but without resistance. After this first treatment phase, Group B (who had received sham treatment) began with EMST treatment, whereas Group A (who already had undergone the EMST treatment) started with maintenance training. After Group B's training with EMST, they also started with maintenance training. Since both groups (A and B) received the same amount of training, they will be referred to as Group A and Group B throughout the manuscript when referring to the group's different protocols, instead of using the terms intervention group and control group.

Speech-language pathologists (SLPs) at each centre assigned eligible participants randomly, i.e. all participants' names were put in a jar and then drawn one on one, and every other was assigned to group A or B. Participants were blinded to treatment allocation, but had received information that they would receive respiratory training with or without resistance. The SLPs who carried out the data collection were not blind to treatment allocation. Additional descriptions of the study protocol are found under Procedures and an overview is found in Figure 1.

The project was approved by The Swedish Ethical Review Authority (DNR: 2019-01402). Written informed consent was obtained from all participants. The study was registered at the Swedish research database Research Web, reference number 270611.

Participants

Patients with PD or MS were recruited via clinics for speech-language therapy at two regional hospitals in the Swedish region Västra Götaland (referred to as Clinic 1 and Clinic 2). The primary inclusion criterion was subjective complaints of feeling out of breath when speaking, and experience of speech and swallowing difficulties, both being a result of the course of the disease. Additional criteria were that patients were in a stable phase of their disease. Exclusion criteria included high blood pressure; respiratory diseases such as asthma or chronic obstructive pulmonary disease; a score <25 on Mini-Mental State Examination (MMSE; Folstein et al. 1975); other neurological diseases or a disease that affects voice function, speech, and/or swallowing; and involvement in other types of physical exercise during the data collection. A further criterion was that the disease severity was scored ≤ 4 on the modified Hoehn & Yahr scale (Goetz et al., 2004) for the participants with PD and ≤ 5 on the Expanded Disability Status Scale (EDSS; Kurtzke, 1983) for patients with MS. Due to problems obtaining the EDSS score during the recruitment process, the cutoff on the EDSS was later adjusted since the individuals with MS who were interested in participating in the study had scores between 6 and 8 on the EDSS. This meant that they had a more severe disease than the original inclusion criterion. Since they fulfilled all other criteria, they were included in the study.

Before inclusion in the study, an assessment of whether patients met the inclusion criteria was carried out. As part of this assessment, their respiratory pressure was tested with a respiratory pressure metre (MicroRPM, MicroMedical, Ltd., Kent, UK) in order to ensure that they fulfilled the requirement of blowing 30 cm H_20, which is the minimum resistance level on EMST150, the device used for training, and that their technique was satisfactory.

A power calculation was performed using Cohen's d ($\alpha = 0.05$) based on the two primary outcome measures: maximum respiratory pressure (MEP) and maximum phonation time (MPT). Guided by published results (e.g. Johansson et al. 2013), MEP was expected to increase from 79.4–94.6 and MPT from 13.04–14.9. From these calculations, a sample size of $n = 3$ based on MEP and $n = 27$ based on MPT was given. Hence the aim was to include 30 participants. According to Cohen (1988), a sample of $n = 16$ in each group would give a 70–80% power to detect effect sizes of > 0.8.

Description of EMST

The device used for training was an EMST150 (Aspire Products, Gainsville, FL, USA), which is a threshold device used for expiratory muscle strength training. EMST150 has a valve that opens when sufficient expiratory pressure is produced. To avoid air leakage, a noseclip is used and each participant could choose between a flanged or rounded mouthpiece. The resistance in EMST150 ranges from 30–150 H_2O, with intervals of 30 H_2O. The training level with EMST150, i.e. the set resistance level, was aimed to correspond to 75% of MEP measured with a respiratory pressure metre. However, since this level of resistance sometimes was hard to reproduce in the EMST150, the resistance was always individually adapted using the EMST150 in order to find an optimal level of resistance. The resistance was adjusted by an SLP once a week during training with EMST,

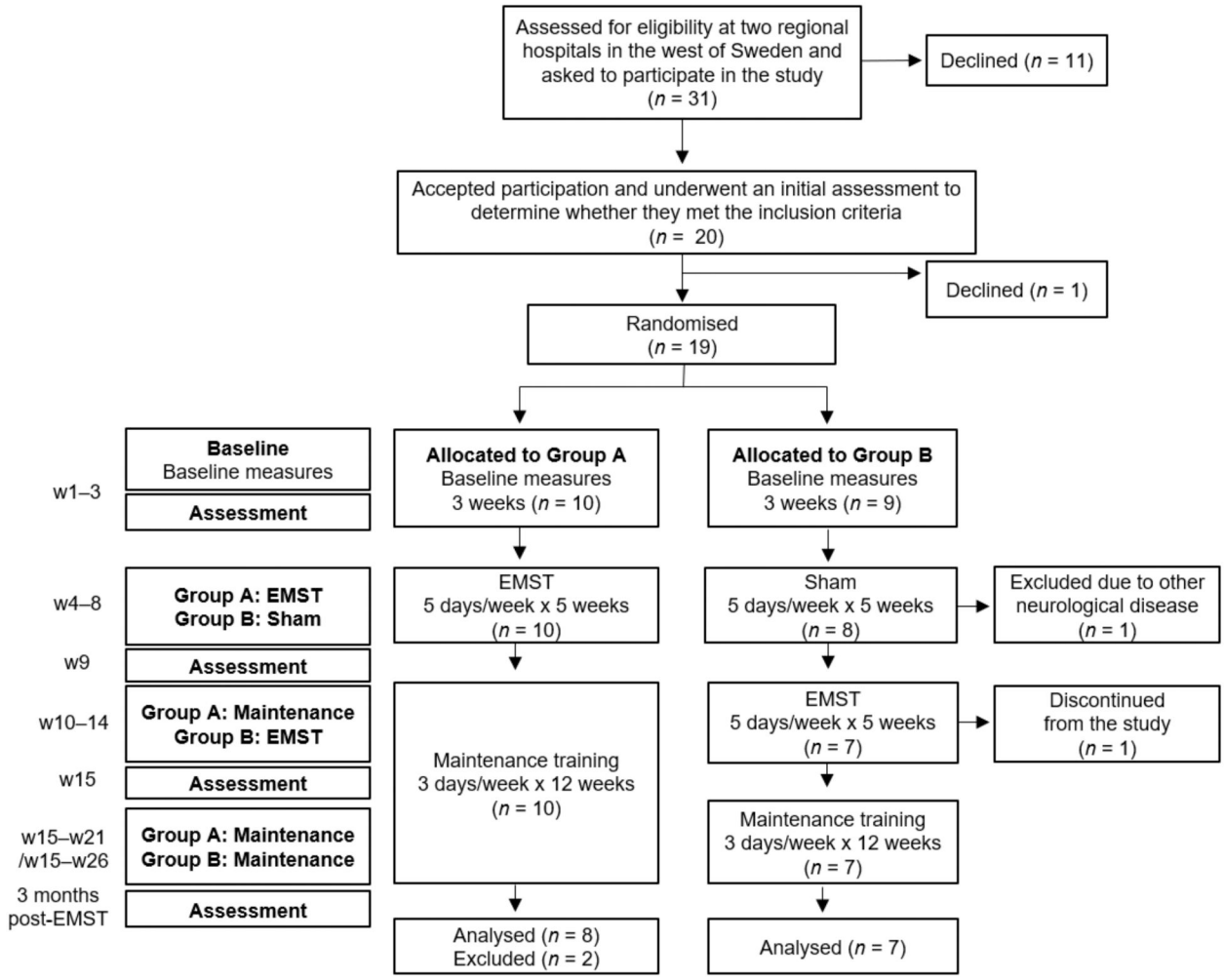

Figure 1. An overview of the study protocol.
Notes: The figure presents an overview of the study protocol, including how many people declined to participate or were excluded at each phase. It further presents how many participants were randomised to each group and the protocol for each treatment arm.

in accordance with the manual supplied by the Swedish retailer (OPIVA Nordic AB). During the maintenance training, the participants continued using the same level of resistance as used at the end of the EMST.

A training session entailed 25 forceful exhalations into the EMST150. After a deep inhalation, the exhalation should be rapid and powerful and last for a few seconds. After each exhalation into the mouthpiece, the patient rested. The exercise was performed five times followed by a short rest of minimum 15–30 seconds, and then this was repeated until participants had performed 25 exhalations with the EMST150. The EMST intervention consisted of a training period lasting 5 weeks with five training sessions per week, continued by 12 weeks of maintenance training where the participant trained 3 days a week. The training was carried out in the participants' homes without the participation of an SLP, but with written instructions. All participants filled in a training diary. Training compliance during EMST as measured by the participants' logs ranged from 80 to 100% in both groups, with an average compliance of 99.6% in the PD group and 98.9% in the MS group. Training compliance during maintenance EMST ranged from 33 to 100% (average 85.2%) in the participants with PD and from 80 to 100% (average 97.7%) in the participants with MS. Data on compliance for the maintenance phase was missing for two participants due to missing logs.

The sham training, which only Group B underwent, entailed exhaling into the rounded mouthpiece separated from the EMST150 device, i.e. without resistance. Apart from the lack of resistance, the same procedure as that for EMST was followed. The participants did not receive information about which group they were randomised to. However, since they were told at the start of the study that they would be randomised to train their respiration with or without resistance, it was possible for the participants to figure out which kind of training they received during weeks 4–8, when Group A started EMST and Group B started sham.

Procedures

Both groups A and B started with a 3 week baseline (weeks 1–3) in order to ensure that the participants

were stable in their outcome measures. During baseline maximum expiratory pressure, maximum phonation time, verbal diadochokinesis, intelligibility, and speech rate was collected once a week. After baseline, Group A trained with EMST and Group B had sham training (weeks 4–8). After this first treatment phase, all participants in both groups underwent the first post-assessment of voice and speech measures (week 9); for an overview of measures see Table I. During weeks 10–14, Group B trained with EMST and Group A had maintenance training, and this was followed by another post-assessment (week 15). Group A then continued with maintenance training (weeks 15–21) and Group B started with maintenance training that continued for 12 weeks (weeks 15–26). After the 12 weeks of maintenance training, the participants underwent a follow-up assessment, i.e. 3 months post-EMST. At the end of the study, both groups had received the same amount of training, i.e. 5 weeks of EMST and 12 weeks of maintenance training.

Besides assessments at baseline and post-assessments after EMST, sham, and maintenance, several of the measures were also collected on a weekly basis; see Figure 1 and Table I. All participants also underwent assessments of their swallowing ability, which will be presented in future publications.

All data collection took place at two speech-pathology units at the regional hospitals included and was carried out by certified SLPs. All measurements were both audio and video recorded. Three SLPs were involved in the data collection at Clinic 1 and two were involved at Clinic 2. The participants with MS and PD had their data collected at different hospitals, except for one participant with PD who went to the same hospital as the participants with MS. Since this participant later was excluded, all participants with the same diagnosis went to the same clinic and consequently met the same SLPs at all assessments. Participants with PD had their testing sessions at similar times in their drug cycles. All SLPs had undergone training in instructing patients to use the EMST device. They had also trained together and developed protocols for a standardised data collection, which in all comprised 3 whole days plus additional meetings.

Primary outcome measures: MEP and MPT

MEP (cm H_2O) was measured using a digital manometer with a flanged mouthpiece (MicroRPM, Respiratory Pressure Metre). A noseclip was used in order to avoid air leakage from the nose. The participant was asked to inhale fully and exhale forcefully as fast as possible. After each exhalation, the participant rested 30–60 seconds or longer if needed. Each value was recorded and the procedure was repeated three to 10 times until the three highest values were within 10% of each other, which then were averaged for the data analysis. This value was also compared with normative values, i.e. predictions of MEP in adults with a flanged mouthpiece from Evans and Whitelaw (2009).

MPT was measured in sustained phonation, where the participant was instructed to take a deep inhalation and sustain a steady /a/ in a comfortable loudness level for as long as possible. The test leader also modelled the task. The task was repeated three times and recorded using a stopwatch. Both mean and maximum values were used for computation, but only the maximum value was used when compared to normative values reported in the Dysarthria Assessment (Hartelius, 2015).

Secondary outcome measures: Diadochokinesis, intelligibility, and speech rate

The DDK task assessed sequential motion rate (SMR) where the participant was instructed to repeat *pa-ta-ka* as quickly and rhythmically as possible. This task was repeated twice and the best performance was used for computation when comparing to normative values reported in the Dysarthria Assessment (Hartelius, 2015).

To assess intelligibility, the participant was asked to read 10 nonsensical sentences including four to five randomly selected content words. The sentences were grammatically correct but semantically impossible, to avoid contextual cues. The percentage of correctly perceived words was used for computation. Participants' scores were also compared to normative values reported in the Dysarthria Assessment (Hartelius, 2015).

Speech rate was calculated in words per minute in a text reading task from the Dysarthria Assessment (Hartelius, 2015). The text used was *The Trapeze Artist* (in Swedish, *Trapetskonstnären*), which is an articulatory complex task designed to reveal articulatory difficulties in mild dysarthria. One participant who had another first language instead read the less complex text *A Severe Fall* (in Swedish, *Ett Svårt Fall*). Normative values for *The Trapeze Artist* and *A Severe Fall* are reported in Hartelius (2015).

Self-reported measures: Self-report form and open questions

To determine whether the intervention had affected the participants' perceived difficulties concerning speech, voice, and communication, the Questionnaire on Acquired Speech Disorders (QASD; Hartelius 2015) was used. This self-report form consists of 30 statements divided into three sections, asking about the individual's perception of their voice and speech (Section A), communicative activity and participation (Section B), and environmental factors affecting communication (Section C). Each statement is rated on an ordinal scale ranging from 0 = *not true at all*, 1 = *sometimes true*, 2 = *mostly true*, to 3 = *always true*. The total score divided by a number

Table I. Overview of timepoints for data collection.

Outcome measures		Timeline																
		Baseline			EMST/sham						EMST/maintenance							
		Group A and B: Collection of baseline measures			Group A: EMST Group B: Sham					Post-assessment	Group A: Maintenance Group B: EMST					Post-assessment	Group A: Maintenance Group B: Maintenance	Post-assessment
		w 1	w 2	w 3	w 4	w 5	w 6	w 7	w 8	w 9	w 10	w 11	w 12	w 13	w 14	w 15	Group A: w 1621 Group B: w 1626	3 months post-EMST
Primary outcome measures:																		
MEP	Mean value of the three highest values within 10%	X	X	X	X	X	X	X	X	X	X	X	X	X	X	X	–	X
MPT	Max and mean of three repetitions	X	X	X	X	X	X	X	X	X	X	X	X	X	X	X	–	X
Secondary outcome measures																		
DDK	Syllables per sec in SMR	X	X	X	X	X	X	X	X	X	X	X	X	X	X	X	–	X
Speech rate	Words per minutes in text reading	X								X								
Intelligibility	% of perceived words	X	X	X	X	X	X	X	X	X	X	X	X	X	X	X	–	X
Self-reported measure																		
QASD	Questionnaire	X								X						X	–	X
Questions about the	intervention						X						X	–	X		Questionnaire	X

MEP = maximum expiratory pressure; MPT = maximum phonation time; DDK = diadochokinetic rate; SMR = sequential motion rate; QASD = Questionnaire on Acquired Speech Disorders (QASD; Hartelius, 2015) self-report form.

Note: After the baseline phase, Group A trained with EMST and Group B trained with sham during 5 weeks (week 4–8). This was followed by a post-assessment for both groups (week 9). Next, Group A started maintenance training and Group B had EMST for 5 weeks (week 10–14). After this both groups underwent a new post-assessment (week 15). Group A continued maintenance training and group B started maintenance training (for weeks see table). After 12 weeks of maintenance training both groups had a new post-assessment, i.e. 3 months after EMST. At this assessment both groups had received the same amount of training, i.e. 5 weeks of intensive training and 12 weeks of maintenance training. X = data was collected.

of items was used for computation, where a higher score is equivalent to a higher degree of perceived difficulties concerning speech, voice, and communication. Besides calculating the total score, two statements from the form that were deemed particularly relevant to the study were chosen for analyses: *it is difficult for me to get enough air when I talk* (A6) and *it is an effort to talk* (A9).

At week 9 (post-EMST or post-sham), week 15 (post-EMST or post 5 weeks of maintenance), and at the 3 month follow-up (post 12 weeks of maintenance EMST) participants were asked to fill in a short questionnaire with the following open-ended questions: (a) How did you experience the intervention?, (b) How has the intervention affected your communication?, and (c) How has the intervention affected your life? The participants answered the questions at home and then returned the questionnaire by post. The participants' answers were coded into three categories: *positive effect* = answers that described a positive effect of the treatment (e.g. "It has helped me with my difficulties when it comes to speech and swallowing" or "It has gotten better. I have more air when I shout"); *marginal/minor effect* = answers that described that the treatment had very little or unclear effects, including answers about gained insight but not clear behaviour change (e.g. "Perhaps. My voice has sometimes become brighter and more even." or "This made me realise I need to train my lungs"); and *no or negative effect* (e.g. "I have not noticed any change" or "dull and monotonous"). The coding was done by two certified SLPs not involved in the data collection. The coders were blinded concerning participants' IDs, the time point when the answers were collected, and what treatment each participant had received. They each coded all answers separately; their agreement calculated using point-to-point concordance was 82.6%. The remaining 17.4%, where the coders disagreed, was discussed together with the first author until a consensus was reached.

Data and statistical analysis

All pre- and post-treatment assessments were analysed by SLPs with clinical expertise in evaluating patients with PD or MS, and who also had trained using the tests and devices used in the project. The majority of the measures were collected offline based on the audio and video recordings, but MPT and MEP were registered during the assessment sessions.

To investigate between-group differences at baseline, Mann–Whitney U-test or Pearson's chi-square were used. To investigate within-group differences over time for the participants with PD or MS, paired comparisons for several samples with Friedman test were used. In those comparisons baseline, post-EMST (i.e. post-assessment week 9 for Group A and post-assessment week 15 for Group B), and post-maintenance EMST were compared. In these calculations, the baseline measures from week 1–3 and measures from week 4, i.e. the week when Group A started EMST and Group B began sham, were averaged to represent baseline performance, whereas the latter points for comparisons were represented by a single data point. Week 4 was included in the mean baseline measures since the measures from that week were collected before the intervention/sham started, hence representing participants' ability pre-intervention. When Friedman test was significant, paired Wilcoxon signed-rank test was used for post-hoc analysis and standardised effect sizes were calculated using Cohen's d_z, a version of Cohen's d adapted for within-group comparisons. The following interpretations of Cohen's d_z was used: Z = −1 – 1 = small difference, Z = 1–2/-2 – −1 = moderate difference, Z = 2–3/−3 – −2 = large difference, and Z > 3 or < −3 = very large difference. Mann–Whitney U was used to examine the differences between EMST and sham. Due to the study's exploratory nature, no adjustment for multiple comparisons was used and the level of significance was set to $p < .05$ for all comparisons. IBM SPSS Statistics version 28 was used for computation.

To complement the statistical analyses, plots visualising how group A and B performed on the different outcome measures over time were constructed. To explore between-group differences, the data were normalised using relevant norms, since group A and B had different proportions of men and women. The plots illustrate the median and the third and first quartile in a time-series design. Non-overlapping data, i.e. if data points in the two time series do not overlap, indicate that there is a difference. In the case of missing values, imputation was done using a mean of the last and the next value in the time series, which is suitable in time-series data. The main reason for imputing data was that approximately 50% of the data in Group A were missing between weeks 10 and 14 (when they began maintenance training), but imputed data were also applied for single missing values in other weeks. The imputed values were only used for the visualisation, not for statistical computation.

To provide an overview of the individual results on MPT, verbal DDK, intelligibility, and speech rate at baseline and post-maintenance EMST, each participant's results were compared to appropriate norms and computed into z-scores. A cutoff at > 1.5 *SD*, which is a common cutoff in a clinical setting for performance, was used as an indication of impairment on these measures. Since there is a lack of what constitutes a clinically relevant change in these measures for these patient groups, a change of ≥1 *SD* was chosen as a cutoff indicating an improvement/decline. MEP values were compared to norms from Evans and Whitelaw (2009) and are presented as a percentage (%) of predicted MEP. Reference values from Evans and Whitelaw (2009) were also used to compare participants' performance in terms of above or under the lower limit of normal (LLN).

Result

Demographics

The study recruited participants during the period March to August 2019. After an initial assessment, 19 patients were included and randomly allocated to Group A ($n = 10$) and Group B ($n = 9$). Due to drop-outs, 17 participants completed the study protocol and two of these were later excluded from the analyses on a group level, see Figure 1 for an overview. The reason for exclusion was epileptic seizures in one participant and a stroke during the course of the study in one participant. Due to these exclusions, all participants with the same diagnosis were tested at the same hospital. There were no significant differences between group A and B with regard to age, sex, diagnosis, smoking history, MMSE score, MEP, or predicted MEP at baseline (Table III). There was a difference in age between the participants with MS and the participants with PD (Table III).

Of the 15 participants included in the analysis, 9 had PD and 6 had MS. The participants with PD had mild or moderate disability according to the modified Hoehn & Yahr scale (Goetz et al., 2004), whereas the participants with MS had moderate or severe disability according to the EDSS (Kurtzke, 1983). There was a variation in both time since diagnosis (PD: $mdn = 5$ years, range 0.5–13 years; MS: $mdn = 12$ years, range 7–20 years) and time since onset of symptoms (PD: $mdn = 6$ years, range 1–18 years; MS: $mdn = 17$ years, range 9–35 years), which is illustrated together with other participant characteristics in Tables I and II. Approximately 50% of the participants with PD were assessed as having mild dysarthria, whereas the vast majority of the participants with MS were assessed as not having dysarthria.

Effects of EMST on the primary and secondary outcome variables for persons with PD and MS: Group level comparisons

In Table IV an overview of the results of all primary and secondary outcome variables at baseline, post-EMST, and after post-maintenance intervention are presented. Both the PD and the MS group increased their MEP significantly, PD group $\chi^2(2) = 8.96$, $p < .001$ and MS $\chi^2(2) = 12.67$, $p = .002$. In the PD group, post hoc analyses revealed an improvement, with a moderate effect size (according to Cohen's d_z) between baseline and post-EMST and between baseline and post-maintenance EMST, and a small decline between post-EMST and post-maintenance. In the MS group, a small improvement was seen between post-EMST and post-maintenance EMST, and a moderate improvement was seen between baseline and post-maintenance. No effect was seen in MPT for any of the groups.

The only secondary outcome measure that differed significantly was DDK, PD group $\chi^2(2) = 8.22$, $p = .016$ and MS group $\chi^2(2) = 10.33$, $p = .006$. For the PD group, post hoc analyses showed a very large improvement between baseline and post-EMST and between baseline and post-maintenance EMST. For the MS group, a very large improvement was seen between baseline and post-EMST, a large improvement was seen between baseline and post-maintenance EMST, and a moderate decline was seen between post-EMST and post-maintenance EMST. Post hoc analyses revealed a large positive effect in DDK for the MS group post-EMST, a very large positive effect post-maintenance EMST, and a moderate decline between post-EMST and post-maintenance EMST.

Effects of EMST on the primary and secondary outcome variables for persons with MS and PD: Individual results

The median increase in MEP (raw scores) from baseline for the PD group was 19% (range 12–28%) post-EMST and 12% (range 0–35%) post-maintenance EMST. For the participants with MS the median increase in MEP was 33% (range 4–113%) post-EMST and 45% (range 6–149%) post-maintenance EMST. None of the participants with PD was assessed as having an MEP under the lower limit of normal (LLN) based on % of predicted values (Evan & Whitelaw, 2009), at baseline, post EMST or post-maintenance EMST. Three participants with MS were assessed as having an MEP under LLN at baseline, two post-EMST, and only one post-maintenance EMST. An overview of the performance on the primary and secondary outcome measures is presented at an individual level in Figure 2 (participants with PD) and Figure 3 (participants with MS). These figures present data on the primary and secondary outcome variables from baseline and post-maintenance EMST, but not post-EMST (descriptive information post-EMST is presented at the group level in Table IV). Of the nine participants with PD, one increased ($\geq$ 1 z-value) and one decreased ($\geq$ 1 z-value) on MPT, five increased their articulatory rate on DDK, two improved their intelligibility and one showed a decline in speech rate. One out of six participants with MS improved their max MPT, four out of six on DDK, one showed a decline and one improved on speech rate.

Comparison between sham and EMST

None of the primary and secondary outcome measures or QASD differed significantly at week 9 when Group A had undergone EMST and Group B had received sham treatment; see Table V. The median improvement in MEP for Group A who had received EMST was 24% (range 4–35%), whereas the median improvement for Group B that had received sham was 7% (range −44–35%). Within-group differences were also examined for group A and B, which showed

Table II. Demographic information and disease characteristics for the participants.

ID	Group	Sex	Age	Time since symptoms onset	Time since disease onset	Disease progression(H&Y max score 5, EDSS max score 10)	Severity of dysarthria	Dysarthria score (max 3)	Previous speech treatment	Percent of predicted maximum expiratory pressure (%)
Participants with PD:										
1P	B	M	61	1	0.5	H&Y 3	Normal	0.02		91%
2P	B	F	71	8	7	H&Y 2.5	Mild	0.9		93%
3P	A	M	82	2	1	H&Y 2.5	Mild	0.7		82%
4P	B	M	67	5	4	H&Y 2	Mild	0.5	LSVT	89%
5P	A	F	67	10	5	H&Y 2	Mild	0.2	LSVT	107%
6P	A	M	79	12	10	H&Y 2	Normal	0.1		115%
7P	B	F	73	3	3	H&Y 1.5	Normal	0.2		129%
8P	A	M	74	6	5	H&Y 2	Normal	0.1		93%
9P	A	M	70	18	13	H&Y 3	Normal	0.1		111%
Participants with MS:										
1M	A	F	63	30	20	EDSS 8	Mild	0.1		51%
2M	A	M	57	9	7	EDSS 6	Normal	0.06		68%
4M	B	F	53	11	9	EDSS 6	Normal	0		140%
5M	A	F	58	15	9	EDSS 7.5	Normal	0.06		179%
6M	B	F	59	35	15	EDSS 6	Normal	0.08		75%
7M	B	F	54	19	17	EDSS 6.5	Normal	0.06		48%

LSVT = Lee Silverman Voice Treatment. *Note*: Disease severity for the PD patients is presented according to the 5-point modified Hoehn and Yahr scale (Goetz et al., 2004) and the Expanded Disability Status Scale (EDSS; Kurtzke, 1983) is used for the patients with MS. For both scales, higher numbers are equated with greater difficulties. The dysarthria score was based on the Swedish Dysarthria Assessment (Hartelius, 2015) where the maximum score of 3 indicates severe dysarthria, scores around 2 indicate moderate dysarthria, scores around 1 indicate mild dysarthria, and scores around 0 indicate normal performance or very subtle difficulties.

Table III. Demographic information and disease characteristics of all participants divided by group.

	Descriptive information divided by groups			Descriptive information divided by diagnosis		
	Group A $n=8$	Group B $n=7$	Sig.	MS ($n=6$)	PD ($n=9$)	Sig.
Age, mdn (range)	68.5 (57–82)	61 (53–73)	$p = .232$	71.5 (61–82)	57.3 (53–63)	$p < .001$
Sex	3 females, 5 males	5 females, 2 males	$p = .189$	3 females, 6 males	5 females, 1 male	$p = .057$
Diagnosis	3 MS/5 PD	3 MS/4 PD	$p = .833$	–	–	
Smoking history	2 ex-smokers, 6 non-smokers	3 ex-smokers, 4 non-smokers	$p = .464$	2 ex-smokers, 7 non-smokers	3 ex-smokers, 3 non-smokers	$p = .264$
MMSE, mdn (range)	29 (25–30)	28 (25–30)	$p = .463$	28 (25–30)	29.5 (25–30)	$p = .456$
MEP baseline, mdn (range)	96 (39–145)	88 (40–120)	$p = .536$	104 (65–129)	73 (39–145)	$p = .328$
Predicted MEP baseline, mdn (range)	100% (51–179%)	91% (48–140%)	$p = .867$	93% (82–129%)	71% (48–179%)	$p = .328$
Previous LSVT	1	1	–	–	2	–

mdn = median; MMSE = Mini-Mental State Examination; MEP = maximum expiratory pressure; LSVT = Lee Silverman Voice Treatment.
Note. Statistical comparisons were done using Mann–Whitney U test (age, MMSE, MEP, predicted MEP) or Pearson's chi-square (sex, diagnosis). The majority of measures are presented with median and range.

Table IV. Effect of expiratory muscle strength on primary and secondary outcome variables, and score on the self-report form Questionnaire on Acquired Speech Disorders (QASD) for the Parkinson's disease and the multiple sclerosis groups.

	Baseline (BL) Mdn (range)	Post-EMST (PE) Mdn (range)	Follow-up, i.e., post-maintenance EMST (PmE) Mdn (range)	Paired comparison *p*-value	Post hoc analyses	Standardised effect size *Z*
PD-group (n =9)						
MEP	104 (65–129)	124 (78–159)	122 (77–142)	**>.001**	BL < PE, BL < PmE, PE > PmE	1.76 1.32 0.51
Mean MPT	10.3 (5.6–25)	10.5 (5.7–35.0)	14.7 (6–36.6)	.169		
Max MPT	12.3 (6.3–28)	12 (6–37)	16 (7–38)	.264		
DDK	5.3 (4.8–6.2)	6.1 (5.4–7.8)	6.1 (5.5–7.1)	**.016**	BL < PE, BL < PmE, PE = PmE	3.18 3.82
Intelligibility	97.8 (60.3–100)	98 (66–100)	96 (64–100)	.247		
Speech rate	113 (44.9–134)	102 (49.5–132)	101.3 (48.6–132.7)	.895		
QASD (max score = 3)	0.6 (0.1–2.1)	0.45 (0.2–2.5)	0.3 (0.2–1.9)	.248		
MS group (n = 6)						
MEP	73 (39–145)	119 (53–151)	125 (63–153)	**.002**	BL < PE, BL < PmE, PE = PmE	0.95 1.29
Mean MPT	12 (5.9–26.1)	16.9 (8.0–31.9)	16.7 (8.9–27)	.513		
Max MPT	13.3 (6.5–28.5)	18.5 (8.9–35.4)	17.3 (10.8–28.9)	.607		
DDK	5.2 (4.5–5.6)	6.3 (5.7–6.8)	6 (5–6.5)	**.006**	BL < PE, BL < PmE, PE > PmE	4.6 2.20 1.70
Intelligibility	99.8 (97.5–100)	100 (97–100)	99 (98–100)	.926		
Speech rate	106.9 (90.5–123.9)	105.6 (94.8–127.2)	102 (94.2–131.0)	.513		
QASD (max score = 3)	1 (0.1–1.8)	0.85 (0.1–1.6)	0.7 (0.0–1.6)	.066		

Nonparametric paired comparisons of several samples using Friedman test with post hoc analysis using Wilcoxon sign rank test. Standardised effect size calculated with Cohen's d_z. Significant values are bolded.
Abbreviations: mdn = median.

that none of the outcome measures changed after 5 weeks of sham, whereas Group A showed significant improvement following EMST on MEP (p =.012), predicted MEP (p =.012), mean MPT (p =.025), max MPT (p =.036), and DDK (p =.012).

To complement these analyses and explore how the outcome variables varied over time, the data distribution was plotted in a series of line charts where both groups are plotted together. The line chart presenting the MEP data is presented in the manuscript (Figure 4), whereas the other line charts can be seen in a supplementary text (Appendix A). The line chart in Figure 4 visualises the raw scores (4a) and normalised values (4b) in order to enable comparisons

between groups. Despite different treatments, a visual inspection of Figure 4(b) showed that there was a large overlap in MEP between the groups for weeks 4–9.

Effects of EMST on the self-report form

Due to missing data for one participant with MS and one participant with PD, the comparison with the self-report form QASD only included five participants with MS and eight with PD. The statistical analysis revealed no significant difference between baseline, post-EMST, and post-maintenance EMST; see Table IV. The MS group's and the PD group's results on the self-report form QASD are visualised in Figure 5.

The participants' answers, at baseline and post-maintenance EMST, to statements A6 (*it is difficult for me to get enough air when I talk*) and A9 (*it is tiring to talk*) are presented in Figures 2 and 3. Post-maintenance EMST, *difficulties getting enough air when talking* decreased in three of six participants with MS and in one with PD. *It is tiring to talk* occurred less frequently in two participants with MS and in five with PD; see Figures 2 and 3.

Experience of EMST and its effect on communication and everyday life

An overview of the analyses of the participant's responses to the open questions can be viewed in Figures 2 and 3. The majority of the participants in both groups expressed a positive experience of the intervention (PD group: 89%, i.e. 8/9 participants post-EMST, and 86%, i.e. 6/7 participants post-maintenance EMST; MS group: 60%, i.e. 3/5 participants post-EMST, and 100%, i.e. 5/5 participants post-maintenance EMST). None of the participants expressed a negative experience of EMST.

Concerning the effect on communication, a larger part of the MS group expressed a positive change following EMST than the PD group (PD group: 33%, i.e. 3/9 participants post-EMST, and 50%, i.e. 3/6 participants post-maintenance EMST; MS group: 80%, i.e. 4/5 participants post-EMST, and 100%, i.e. 5/5 participants post-maintenance EMST). The MS group also reported a higher proportion of positive changes in everyday life than the PD group (PD group: 60%, i.e. 3/5 participants post-EMST, and 88%, i.e. 7/8 participants post-maintenance EMST; MS group: 100%, i.e. 5/5 participants post-EMST, and 80%, i.e. 4/5 participants post-maintenance EMST).

Although not presented in Figures 2 and 3, Group B also answered the questions post-sham. Three out of five participants (60%) reported a positive experience of the intervention, one out of six (17%) expressed a positive effect on communication, and none expressed a positive effect on life in general.

Discussion

This was an exploratory study examining how EMST affects respiration, voice, and speech in persons with PD or MS. In line with previous studies, both the PD group and the MS group improved their expiratory muscle strength following EMST (e.g. Chiara et al., 2007; Darling-White and Huber 2017; Gosselink et al., 1999; Reyes et al. 2020; Srp et al., 2021). This improvement was still evident at the 3 month follow-up after the 12 weeks of maintenance training. The group with MS did in fact increase their MEP additionally between the end of EMST and post-maintenance EMST, whereas the group with PD showed a small decline, but that decline was not significant. The use of a maintenance program has been suggested by previous studies, which reported a decline in MEP after a period of no training in persons with PD (Troche et al., 2014) or MS (Srp et al., 2021), and to our knowledge this is the first study that included a maintenance phase after EMST. Due to the progressive nature of the disease, sustained or maintenance training is recommended for persons with PD following a program of intensive physical exercise (Mak et al., 2017). A similar rationale for persons with MS seems reasonable, given the slow progressive nature of the disease.

The rationale for including both MS and PD in the same study was that respiration, voice, and speech are affected in both diseases and that both diagnoses are highly prevalent in SLP clinics. Despite the fact that there are differences in neuropathology behind the respiratory symptoms in PD and MS, previous studies have found promising results using EMST to improve expiratory muscle strength in both MS (e.g. Chiara et al., 2007; Gosselink et al., 2000; Smeltzer et al., 1996) and PD (e.g. Darling-White & Huber, 2017; Sapienza et al., 2011; Reyes et al., 2020). EMST is a set intervention and treatment regime developed over the years, and we consider that it was a legitimate aim to test this intervention on both groups. Furthermore, the study did not have an explicit aim to determine whether EMST was more beneficial in one group or the other. It would have been preferable if we also could have compared the groups, but due to differences between the participants with MS and PD, this was not deemed possible. The participants with MS had a more severe disease than the participants with PD, and there was also an age difference between the two groups.

Both the PD and MS group had a median MEP value under that predicted, but this was more evident for the MS group (*mdn* % of predicted MEP for the PD group = 93, and MS group = 71), and only participants in the MS group (three out of six) performed below the lower limit of normal according to the reference values suggested by Evans and Whitelaw (2009). A relationship between MEP and disease severity has been reported in both PD patients (Wang et al., 2014) and MS patients (Gosselink et al.,

Demographics				Raw-scores and normalised scores for outcome measure at baseline (BL) and post-maintenance EMST (PmE)										Questionnaire on Acquired Speech Disorders (QASD)				Self-reported change					
ID	Sex	Age	H&Y	MEP (% of predicted MEP)		MPT, max in s (z-value)		DDK syllables/sec (z-value)		Intelligibility % in sentences (z-value)		Speech rate words/min (z-value)		Problems getting air talking		Tiring to speak		Experience of EMST		Effect on commun-ication		Effect on everyday life	
				BL	PmE	BL	PmE	BL	PmE	BL	PmE	BL	PmE	BL	PmE	BL	PmE	PE	PmE	PE	PmE	PE	PmE
1P	M	61	3	113 (91)	124 (101)	13 (-0.9)	18 (-0.5)	6.5 (0.7)	6.1 (0.3)	98.3 (0.6)	96 (-0.3)	118.5 (-1.1)	121 (-0.9)	No	No	No	No	P	M	N	N	- -	P
2P	F	71	2.5	65 (93)	77 (111)	9 (-1.2)	8 (-1.3)	5.6 (-0.2)	6.5 (0.7)	60.3 (-13.2)	64 (-11.9)	44.9 (-6.4)	48.6 (-6.1)	Often	Often	Often	ST	P	P	P	P	P	P
3P	M	82	2.5	87 (82)	93 (88)	6.3 (-1.4)	18 (-0.5)	5.2 (-0.6)	6.0 (0.2)	86.8 (-3.7)	95 (-0.7)	67 (-2.9)	61.6 (-3.2)	Often	Often	ST	Often	P	-	P	-	-	-
4P	M	67	2	105 (89)	122 (103)	9.5 (-1.1)	10 (-1.1)	5.2 (-0.6)	6.5 (0.7)	69.8 (-9.8)	72 (-9.0)	60 (-3.4)	66.3 (-3.2)	ST	No	ST	No	P	P	M	M	M	P
5P	F	67	2	78 (107)	94 (128)	28 (1.3)	16 (-0.3)	5.3 (-0.5)	5.9 (0.1)	97.8 (-0.1)	100 (0.7)	113 (0.1)	101.3 (-1.0)	ST	Often	ST	Often	M	-	M	- -	P	P
6P	M	79	2	124 (115)	124 (115)	24.5 (0.0)	38 (1.1)	5.3 (-0.5)	5.5 (-0.3)	100 (1.1)	98 (0.4)	134 (0.6)	132.7 (0.6)	ST	ST	Always	Often	P	P	N	- -	-	M
7P	F	73	1.5	88 (129)	99 (145)	12.3 (-0.7)	13 (-0.6)	6.3 (0.5)	7.1 (1.2)	98.3 (0.1)	100 (0.7)	114.8 (0.3)	119 (0.7)	ST	ST	ST	ST	P	P	M	M	M	P
8P	M	74	2	104 (93)	141 (125)	6.9 (-1.4)	7 (-1.3)	4.9 (-1.0)	6.0 (0.2)	99.5 (0.9)	100 (1.1)	117 (-0.3)	109.5 (-0.7)	ST	ST	ST	No	P	P	M	P	-	P
9P	M	70	3	129 (111)	142 (122)	20.9 (-0.3)	31 (0.5)	5.1 (-0.7)	6.3 (0.5)	93.3 (-1.3)	94 (-1.1)	102 (-1.1)	100 (-1.2)	ST	ST	Often	No	P	P	P	P	P	P
			Median:	*104 (93)*	*122 (115)*	*12.3 (-0.9)*	*16.0 (-0.5)*	*5.3 (-0.6)*	*6.1 (0.3)*	*97.8 (-0.1)*	*96.0 -0.3*	*113 (-1.1)*	*101.3 (-1.0)*										
			Below normal:	0	0	0	0	0	0	3	2	3	3										
			Improved/declined:			1 improved 1 declined		3 improved		2 improved		1 declined		1 improved 1 declined		5 improved 2 decline		89%	86%	33%	50%	60%	88%

Colour coding MEP			Colour coding z-values						Colour coding QASD					Colour coding Self-reported change					
	>LLN	<LLN		> 2	1 – 1.9	1 – -0.9	-1 – -1.9	< -2		No	ST	Often	Always		Positive	Marginal	No/ negative	Missing	Irrelevant

Figure 2. An overview of the PD participants' performance on primary and secondary outcome measures at baseline (BL) and post-maintenance EMST (PmE), presented together with self-reported measures (QASD at BL and PmE and open questions at PE, i.e. post-EMST and PmE).

Notes: The figure visualises each participant's performance on MEP, max MP T, DDK, intelligibility, and speech rate. Both raw scores and normalised values are presented. Besides individual results, the figure also presents the group median, how many participants performed below normal, and how many improved or showed a decline, i.e. changed 1 standard deviation/z-score. The answers to two of the questions from QASD (A6 and A9) are presented with the four scale steps: no, often, sometimes (ST), and always. A change in one step on the scale was considered as an improvement or decline on QASD. The analyses of the participants' answers to the open questions are reported in the categories: positive (P), marginal (M), no or negative (N). Missing data are marked with -, and answers containing irrelevant information are marked with - -. Below each column the percentage of how many participants reported a positive experience/effect is reported. In these percentages, missing data and responses containing irrelevant information are excluded. The figure also includes colour coding and the keys to the colour coding can be seen at the bottom of the figure.

Demographics				Raw-scores and normalised scores for outcome measure at baseline (BL) and post-maintenance EMST (PmE)										Questionnaire on Acquired Speech Disorders (QASD)				Self-reported change					
ID	Sex	Age	EDSS	MEP/ % of predicted MEP		MPT, max in s (z-value)		DDK syllables/sec (z-value)		Intelligibility % in sentences (z-value)		Speech rate words/min (z-value)		Problems getting air talking		Tiring to speak		Experience of EMST		Effect on commun-ication		Effect on everyday life	
				BL	PmE	BL	PmE	BL	PmE	BL	PmE	BL	PmE	BL	PmE	BL	PmE	PE	PmE	PE	PmE	PE	PmE
1S	F	63	8	39 51	64 83	7.9 (-1.3)	10.8 (-0.9)	4.6 (-1.2)	5.3 (-0.5)	99.5 (0.6)	98 (0.0)	90.5 (-2.6)	99 (-1.9)	ST	No	ST	ST	M	P	P	P	P	P
2S	M	57	6	86 68	116 92	28.5 (0.3)	26.8 (0.2)	5.5 (-0.4)	6.0 (0.2)	97.5 (0.3)	98 (0.5)	123.9 (-0.7)	131 (-0.1)	Often	ST	ST	ST	-	P	-	P	-	P
4S	F	53	6	120 140	134 157	22.9 (0.6)	28.9 (1.4)	4.7 (-1.1)	5.0 (-0.8)	100 (0.8)	100 (0.8)	109 (-1.2)	94.2 (-2.3)	ST	-	ST	No	P	P	P	P	P	P
5S	F	58	7.5	145 179	153 189	13.6 (-0.6)	16.4 (-0.2)	5.5 (-0.3)	6.2 (0.4)	100 (0.8)	100 (0.8)	108 (-1.1)	130.8 (0.8)	No	No	No		M	-	P	-	P	-
6S	F	59	6	60 75	150 186	6.5 (-1.5)	18.2 (0.0)	5.6 (-0.2)	6.5 (0.7)	100 0.8	98 (0.5)	101.3 (-2.5)	102 (-2.5)	ST	No	ST	No	P	P	P	P	P	P
7S	F	54	6.5	40 48	63 74	13.0 (-0.7)	12.4 (-0.7)	5.5 (-0.4)	6.0 (0.2)	99 (0.4)	100 (0.8)	105.8 (-1.3)	102 (-1.7)	ST	No	N	N	P	P	N	P	P	M
			Median:	*73 (71)*	*125 (124)*	*13.3 (-0.6)*	*17.3 (-0.1)*	*5.5 (-0.4)*	*6.0 (0.2)*	*99.8 (0.7)*	*99.0 (0.6)*	*106.9 (-1.3)*	*102 (-1.8)*										
			Below normal:	3	1	0	0	0	0	3	2	3	3										
			Improved/declined:			1 improved		0 improved		0 improved		1 declined 1 improved		4 improved		2 improved		60%	100%	80%	100%	100%	80%

Colour coding MEP			Colour coding z-values						Colour coding QASD					Colour coding Self-reported change					
	>LLN	<LLN		> 2	1 – 1.9	1 – -0.9	-1 – -1.9	< -2		No	ST	Often	Always		Positive	Marginal	No/ negative	Missing	Irrelevant

Figure 3. An overview of the MS participants' performance on primary and secondary outcome measures at baseline (BL) and post-maintenance EMST (PmE), presented together with self-reported measures (QASD at BL and PmE and open questions at PE, i.e. post-EMST and PmE).

Notes: The figure visualises each participant's performance on MEP, max MP T, DDK, intelligibility, and speech rate. Both raw scores and normalised values are presented. Besides individual results, the figure also presents the group median, how many participants performed below normal, and how many improved or declined, i.e. changed 1 standard deviation/z-score. The answers to two of the questions from QASD (A6 and A9) are presented with the four scale steps: no, often, sometimes (ST), and always. A change in one step on the scale was considered as an improvement or decline in QASD. The analyses of the participants' answers to the open questions are reported in the categories: positive (P), marginal (M), no or negative (N). Missing data are marked with -, and answers containing irrelevant information are marked with - -. Below each column the percentage of how many participants reported a positive experience/effect is reported. In these percentages, missing data and responses containing irrelevant information are excluded. The figure also includes colour coding and the keys to the colour coding can be seen at the bottom of the figure.

2000). It is possible that differences in MEP might partly be related to differences in disease severity. However, this was not obvious in the MS group where three of the participants had an MEP above the predicted level despite a moderate to severe level of disability.

Table V. Between- and within-group comparison between baseline and post-expiratory muscle strength training or sham.

	Between-group comparisons						Within-group comparisons					
	Baseline (BL)			Post-EMST vs. sham			Group A			Group B		
	Group A Mdn (range)	Group B Mdn (range)	*p* value	Group A (post-EMST) Mdn (range)	Group B (post-sham) Mdn (range)	*p* value	BL Mdn (range)	Post-EMST Mdn (range)	*p* value	BL Mdn (range)	Post-sham Mdn (range)	*p* value
MEP	96 (39–145)	88 (40–120)	.536	121 (53–159)	90 (23–129)	.072	96 (39–145)	121 (53–159)	**.012**	88 (40–120)	90 (23–129)	.499
% of predicted MEP	100 (51–179)	91 (48–140)	.867	124 (69–187)	109 (27–142)	.613	100 (51–179)	124 (69–187)	**.012**	91 (48–140)	109 (27–142)	.499
Max MPT	17 (6.3–28.5)	10.3 (6.5–22.9)	.463	21 (6–37)	12.1 (8–25.4)	.281	17 (6.3–28.5)	21 (6–37)	**.036**	10.3 (6.5–22.9)	12.1 (8–25.4)	.237
Mean MPT	17.2 (5.6–26.1)	12.5 (5.9–22.8)	.536	19 (5.7–35)	11.7 (6.7–22.8)	.281	17.2 (5.6–26.1)	19 (5.7–35)	**.025**	12.5 (5.9–22.8)	11.7 (6.7–22.8)	.345
DDK	5.1 (4.7–5.5)	5.3 (4.5–6.2)	.281	6 (5.4–7.2)	6.1 (4.8–7.5)	.463	5.1 (4.7–5.5)	6 (5.4–7.2)	**.012**	5.3 (4.5–6.2)	6.1 (4.8–7.5)	.063
Intelligibility	98 (86.8–100)	97 (60.3–100)	.867	99 (97–100)	97 (61–100)	.189	98 (86.8–100)	99 (97–100)	.612	97 (60.3–100)	97 (61–100)	.753
Speech rate	110.5 (67–134)	105.8 (44.9–118.5)	.397	114.2 (57–132)	104 (52–117.8)	.232	110.5 (67–134)	114.2 (57–132)	.674	105.8 (44.9–118.5)	104 (52–117.8)	.866
QASD	0.8 (0.1–1.2)	1.0 (0.1–2.1)	.336	0.5 (0.3–1.5)	1.2 (0.1–1.5)	.282	0.8 (0.1–1.2)	0.5 (0.3–1.5)	.609	1 (0.1–2.1)	1.2 (0.1–1.5)	.279

mdn = median; BL = baseline; QASD = Questionnaire on Acquired Speech Disorders (QASD; Hartelius 2015).
Note. Nonparametric comparisons of independent samples using Mann–Whitney and paired sample comparisons using Wilcoxon signed-rank test. Significant values are bolded.

Modest effects on measures of voice and speech

Besides the improvement in MEP, the only other outcome measure that improved post-EMST was DDK. Both the MS and the PD groups had an improved performance in DDK post-EMST and this effect was seen after 3 months of maintenance training. If the improvement in DDK was due to the EMST or a practice effect due to repeated testing is not clear. The only other study to our knowledge to investigate the effects of EMST on DDK is Johansson et al. (2013) who used it as a control variable and did not find a change following EMST. DDK is often used to assess the speed and precision of articulatory movements, and it is not obvious that improved expiratory pressure results in a better performance on a DDK task. Another possible explanation for the improvement is a learning effect. Since the task was performed once a week, between week 1 and 15, the participants had probably also become more familiar with the task which might partly explain their improvement in DDK. Then again, the improvement in DDK was not seen following sham. Furthermore, the improved performance in DDK was preserved after the maintenance phase when Group A only was tested in the first weeks.

As mentioned, no significant change was seen in MPT, intelligibility, or speech rate post-EMST for either patient group. To our knowledge, this is the first study to include intelligibility as an outcome measure, but some studies did include maximum phonation time and speech rate. In line with our results, Chiara et al. (2007) did not find any effect on maximum phonation time or speech rate after EMST. They suggested that uncoordinated laryngeal and expiratory musculature activity might explain the lack of improvement in maximum phonation time, despite the improvement in MEP. The participants increased their speech rate (measured in a reading task) only after a detraining phase, i.e. no training, which they attributed to a learning effect. With regard to the PD population, Reyes et al. (2020) found that EMST improved maximum phonation time in participants with mild to moderate PD. The aim of their study was to improve voice intensity and they saw an improvement in sound pressure level and subglottic pressure measured in plosives. The present study did not include voice intensity as an outcome measure. Only two other studies have explored the effects of EMST on voice and speech, reporting results on an individual level. Johansson et al. (2013) used a single-subject design to explore the effects on voice and speech in persons with MS. All five increased their voice intensity, and three out of five increased their maximum phonation time, while four out of five increased their intensity during maximum phonation time and three out of five increased their intensity during reading. In Darling-White & Huber (2017) only two out of nine participants with PD improved

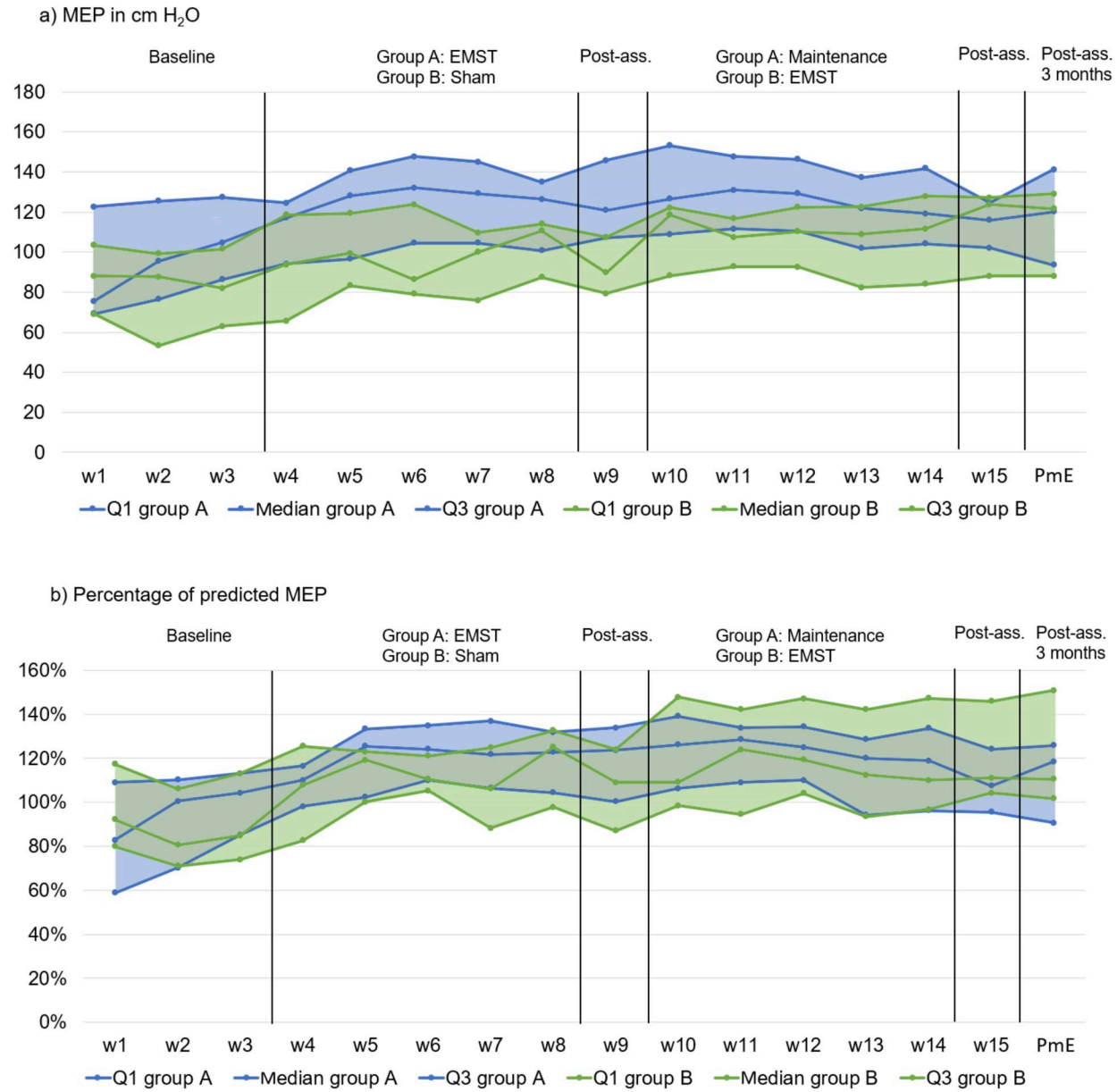

Figure 4. Line chart visualising group A and B's performance on maximum expiratory pressure.
Notes: Line charts visualising Group A (in blue) and Group B's performance (in green) on MEP from week 1 until the end of the protocol post-maintenance EMST -intervention. The middle line represents the median, whereas the upper and lower lines represent the third and first quartile. Visualises MEP in cm H2O, whereas in 4(b) percentages of predicted MEP are plotted in order to compare the scores to norms and illustrate the groups' overlap. Post ass: post assessment.

in their voice intensity and two showed a decline. Furthermore, four participants increased and one decreased their utterance length, i.e. the number of syllables produced during one breath.

One possible explanation as to why only modest improvements were seen in voice and speech in the present study might be a ceiling effect. Most participants in both groups performed within the normal range on all the voice and speech measures. Additionally, despite those subjective complaints of feeling out of breath when speaking was an inclusion criterion, the majority of the participants rated this as only occurring *sometimes* when the same question was posed to them in the self-report form at baseline testing; two participants reported that this problem had not occurred at all. In summary, if the participants included did not in fact have marked difficulties in voice and speech areas, this might explain why only modest improvements were seen. However, three participants with PD had affected intelligibility, and two out of these improved their intelligibility at the sentence level post-EMST.

Subjective reports of EMST

No change was seen in the level of self-reported difficulties in voice, speech, and communication measured with the self-report form QASD. Chiara et al. (2007), in their study of persons with MS, used a questionnaire on voice-related quality of life that

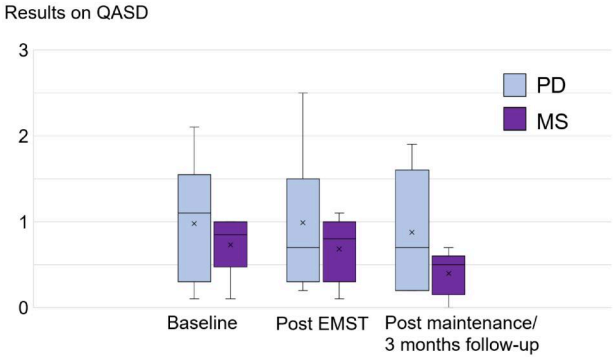

Figure 5. Results on Questionnaire on Acquired Speech Disorders (QASD; self-report form on acquired speech deficits) for participants with Parkinson's disease and multiple sclerosis.
Notes: Boxplots illustrating results on the self-report form Questionnaire on Acquired Speech Disorders (QASD) at baseline, post- EMST, and post-maintenance EMST. Due to missing data, only five participants with MS (blue boxes to the left) and 8 with PD (purple boxes to the right) are included in the plotted data. The QASD score ranges from 0 to 3, where a lower value means fewer difficulties.

partly covered the same areas as asked about in QASD, and did not find any change post-EMST either. As they discussed, there might be several reasons for not finding a change, including that EMST might not have an impact on all of the areas covered by the questionnaire used in the present study. To capture the participants' own experiences, open questions concerning the effects on communication and everyday life were included. The results showed that a majority of the participants with MS reported a positive effect on the communication of EMST. The corresponding number for the PD group was only a third. The reports of positive effects on communication in persons with MS are in concordance with Johansson et al. (2013). In their single-subject study with five persons with MS, all reported a positive experience of EMST. Furthermore, three out of five reported improved respiration and four out of five noted that the air lasted longer when talking, that they could speak in longer phrases, and that communication was better. The authors discussed how the positive comments made by the participants might be partly explained by increased general ventilation due to increased MEP. The research on how respiration is linked to quality of life in persons with PD and MS is scarce. A recent study found a relationship between respiratory muscle strength and aspects of quality of life in persons with MS (Balkan & Salci, 2020), a finding that highlights the need to include a measure of wellbeing when examining the effects of EMST. Another factor that might explain the positive reports by the participants is the importance of engaging in intensive rehabilitative training when suffering from a progressive neurological disease. This was also mentioned in reply to the open questions, where some expressed how they enjoyed the repeated testing, including meeting engaged healthcare personnel as well as following progress. Since there are some uncertainties regarding to what degree respiratory dysfunction impacts everyday life, further qualitative research is needed. This was also concluded in a recent review on respiratory training in Parkinson's disease by van de Wetering-van Dongen and colleagues (2020), highlighting the great difficulty in knowing if better performance on MEP leads to meaningful improvements in real life.

Methodological considerations

This study has several limitations. Most evident is the small sample size. There are studies that found effects of EMST despite a small sample (e.g. Chiara et al., 2007; Reyes et al., 2020). However, it is not clear if our sample size was sufficient to detect differences in the chosen outcome measures since the study not could reach the sample size estimated by the initial power calculation ($n = 16$ in each group). A longer recruitment period would have been preferable, but for practical and economic reasons the recruitment period only lasted for 6 months. A larger sample size would also allow for randomisation balanced for additional factors such as age, gender, and disease severity.

Another factor that might have influenced the result was that the inclusion criteria were based on self-perceived difficulties and not a cutoff on a specific test. All participants experienced problems with regard to respiration, voice, and speech which, together with swallowing difficulties that are studied separately, were inclusion criteria These criteria were chosen based on the fact that subjective difficulties are clinically important and that those who have self-perceived difficulties often are motivated to train and improve those functions. But if the subjective difficulties are too subtle or mild to be reflected in the objective measures of voice, speech, or respiration, it might be difficult to detect any measurable changes post-training. From our data, no clear pattern has emerged that indicates that those with more severe problems improved more than those with an MEP closer to the expected. Hence, this is an important issue to explore in future studies.

The study had a sham-controlled design to investigate a placebo effect. Other studies investigating the effects of EMST on swallowing in persons with PD (Troche et al., 2010) and respiration and swallowing in amyotrophic lateral sclerosis (Plowman et al., 2019) have found that MEP, as well as other outcome measures, can also improve following sham. This highlights the need to control for a placebo effect when participants are undergoing intensive rehabilitation and repeating the same test battery several times. It should thus be noted that, despite small improvements following sham, the studies mentioned above also disclosed significant differences between sham and EMST. In the present study, no differences were seen when comparing sham (Group B) and EMST (Group A), but the within-group analyses differed for

the groups between baseline and post-treatment. An improvement was seen in MEP, MPT, and DDK, for the group who received EMST, whereas no change was seen for the group who received sham. In fact, the group who received sham only improved their MEP by 3% post-treatment compared to baseline. Despite these within-group differences indicating an effect of EMST, the evidence for a specific effect of EMST would have been stronger if a between-group difference was seen at the post-assessment week 9.

Another methodological consideration is repeated testing, which entails a risk of practice effects and have already been discussed with reference to the improvement in DDK. All primary and secondary measures, except speech rate, were collected once a week for 16 weeks in a row. However, if there had been a general practice effect, we would have expected the sham group also to have improved significantly, but this was not seen. This was evident for MEP where Group B did not improve as much as Group A when comparing baseline and performance at week 9, when Group A had received EMST and Group B had received sham (median improvement for those who had received EMST was 24% and median improvement for those who had received sham was 7%). A second aim of including a multiple baseline was that when the actual intervention started, the participants were already familiar with the tests, having performed them for 4 weeks in a row (i.e. pre-testing and baseline). Both groups also had a gap in their testing in the maintenance phase for seven (Group A) or 12 weeks (Group B), before the final testing post-12 weeks of maintenance, so the risk of a practice effect for the comparison between baseline and maintenance EMST appears lower.

Conclusion and clinical implications

To summarise, although the results of this study support previous evidence that EMST has positive effects on expiratory pressure in persons with PD or MS, the evidence for effects on voice and speech remains small. Even though the latter finding can be related to the present study's small sample, the analysis on an individual level showed that few participants improved on the tests of voice and speech, except for the DDK task. Since there is uncertainty over whether the improvement in DDK is an effect of EMST or a practice effect due to repeated testing, this result remains uncertain. Our interpretation of the self-reported measures was that despite its length, the intervention was well tolerated. The participants with MS had a higher proportion of positive reports of the effect on communication and life in general than the participants with PD, which might be linked to a greater improvement in MEP in the MS group. But since these findings are based on a small sample, the results should be interpreted with caution. Besides improvements in MEP, EMST can have effects on measures of voice and speech for patients with PD (Reyes el., 2020) or MS (Johansson et al., 2013). We suggest that outcome measures should be chosen with great care when evaluating EMST in the clinic. Several questions are still unanswered when it comes to the effects of EMST and for which patient groups it is beneficial. Since subjective reports of the intervention and its effects on communication were predominantly positive, in-depth research, preferably using a qualitative approach, is warranted with the aim of both gaining insight into subjective experiences and selecting appropriate outcome measures. Further research is also needed that includes larger groups of participants with various degrees of disease severity, and examining different protocols for EMST and maintenance EMST.

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